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## Journal of Pediatric Surgery Case Reports

journal homepage: [www.jpascasereports.com](http://www.jpascasereports.com)

## Endoscopic membranotomy of a tubular type esophageal duplication cyst performed on a 5 year old child

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## ARTICLE INFO

## Article history:

Received 24 August 2016

Received in revised form

4 October 2016

Accepted 7 October 2016

Available online 10 October 2016

## ABSTRACT

Duplications of the gastrointestinal tract are rare malformations with an incidence of one in 4500 [1]–12500 [2]. Esophageal duplication cysts represent about 10% of all foregut duplications and are classified as cystic or tubular type duplications [3]. The tubular type esophageal duplication cyst is a rare subgroup of the esophageal duplication cysts seen in only 5%–10% of all cases [4]. We report about a 5-year-old boy with such a tubular type esophageal duplication cyst presenting with dysphagia and malnutrition. To our knowledge this is the first report about an endoscopic resection of an intraluminal bridge using an insulated scissors-type knife SB knife (SUMITOMO BAKELITE CO., Tokyo, Japan). The postoperative course was uneventful and the patient could be discharged only one day after surgery. The follow-up endoscopy three month later revealed no evidence of residuals of the duplication or remarkable stricture. We conclude that the endoscopic resection of tubular type esophageal duplication cysts is a save and preferable therapeutic option also in children.

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## 1. Introduction

Duplications of the gastrointestinal tract are rare malformations with an incidence of one in 4500 [1]–2500 [2]. Esophageal duplication cyst is a congenital malformation of the posterior primitive foregut and represents about 10% of all foregut duplications [3]. The clinical presentation depends on the localization of the cyst and can range from dysphagia, regurgitation, tachypnea, chest pain, and other respiratory symptoms until the lack of symptoms. If patients present with clinical symptoms, surgical resection of the esophageal duplication cyst is the advocated therapeutic option. Depending on the localization and size of the cyst different approaches are reported like thoracotomy, thoracoscopy or transhiatal laparoscopy. The endoscopic resection is an upcoming treatment option in tubular type esophageal duplication cysts. In literature there is only one case in children reported, that was treated by endoscopic resection [6].

## 2. Case

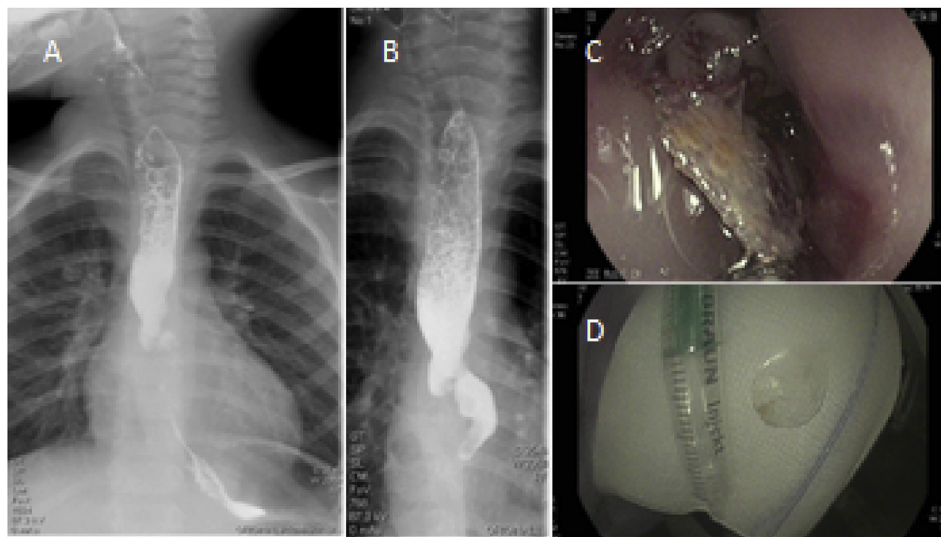
We report about a 5-year-old boy suffering from dysphagia, respiratory distress and recurrent regurgitation persisting for over the last two years without weight loss but slightly malnutrition. The symptoms started at the age of one and a half years after an accidental ingestion of a piece of confetti during a birthday party. Afterwards, the boy has had difficulty in swallowing solid food for over one year. Meanwhile the boy has started to regurgitate even fluids and had a gestational age of 3 to 3 ½ years. The physical examination was normal as well as laboratory values, except the C-reactive protein which was slightly elevated to 1.1 mg/dl. The examinations showed no evidence for congenital abnormalities. The abdominal ultrasound revealed normal anatomic constitution of all organs.

For further diagnostic an esophagram was made showing a stenosis of the distal esophagus (Fig. 1A and B). A CT scan of the chest validated the stenosis and showed reactive lymphatic tissue, with a small hypodense structure dorsal of the esophagus at the level of the aortic arch representing mostly compatible with a diverticle of the esophagus.

A constriction of the esophagus caused by a foreign body granuloma was found at the endoscopic examination of the upper gastrointestinal tract (Fig. 1C). After removing the foreign body

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**Fig. 1.** A, B: Esophagram at initial presentation; C: Initial endoscopy showing the foreign body granuloma; D: removed foreign body.

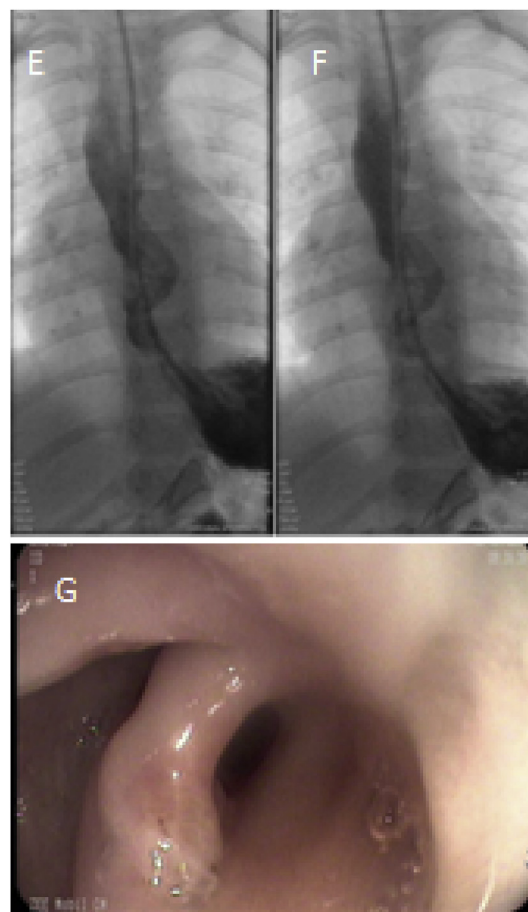
(Fig. 1D), which was compatible with a piece of confetti, the esophagus presented with a double lumen (Fig. 2E and F). We performed another esophagram three days after the endoscopy to verify the diagnosis. The esophagus was intubated with a nasogastric tube and contrast media was applied. In fact the distal part of the esophagus presented with a double lumen. The diagnosis of a rare tubular esophageal duplication was confirmed (Fig. 2G).

Because of the high concern that a surgical resection and anastomosis of the esophageal wall would cause a postoperative stenosis, the decision was made to unify the two esophageal lumina by incising the membrane. This was performed with an insulated scissors- type knife SB knife (SUMITOMO BAKELITE CO., Tokyo, Japan). Therefore we performed an esophagoscopy using a normal gastroscope with a working channel for the insertion of the SB knife. The mucosal bridge was identified at the distal part of the esophagus and removed by cutting and cautery with the SB knife. The whole procedure was performed without any complications in general anesthesia. Cefuroxim was supplied intravenously as a perioperative single shot antibiotic prophylaxis. The postoperative course was uneventful. The oral nutrition could be started immediately after operation with fluids and in the next morning with solid food. The patient could be discharged at the first postoperative day. In the follow-up 3 month after intervention a control esophagogastroscope was carried out. The endoscopy revealed no evidence of residuals of the duplication or remarkable stricture. In the follow-up 18 months later the patient didn't have any symptoms anymore and thrive very well. We recommend a yearly follow-up in case of lacking symptoms.

### 3. Discussion

Esophageal duplication cysts are a rare entity representing about 10% of all foregut duplications, whereas the tubular type is seen in 5%–10% of all cases of esophageal duplication cysts [4]. Their symptoms frequently appear in the first two years of life but they can also incidentally be discovered even in adulthood [7]. Most esophageal duplications are intramural, cystic and non-communicating [5]. Upper esophageal cysts usually become symptomatic much earlier than the lower esophageal cysts. Those patients present generally before the age of 2 years with tachypnea,

stridor, cough or difficulty feeding [7,8]. A duplication cyst in the mid or lower esophagus can be asymptomatic until the cyst becomes infected or inflamed due to retention of esophageal contents



**Fig. 2.** E, F: Esophagram after removing the foreign body; G: Mucosal bridge between tubular type esophageal duplication cyst and the real lumen.

[9]. In the presented case clinical symptoms appeared slightly after ingestion of a foreign body causing a constriction of the esophagus at the level of the tubular esophageal duplication cyst leading to dysphagia and recurrent regurgitation with malnutrition of the child.

The exact genesis of an esophageal duplication cyst is not really understood. But one theory is that the esophageal duplication cyst results from a defect in the tubulation (vacuolization) of the esophagus, occurring in the 6th week of embryonic life [10]. They can be associated with other congenital anomalies, such as small intestinal duplication, esophageal atresia distal to the duplication, tracheoesophageal fistulas, and spinal abnormalities, including scoliosis, hemi vertebrae, and fusion [11,14].

The endoscopic resection of esophageal duplication cysts has increased in adult surgery in the last years because of its minimal invasiveness [12,13]. There are two cases of duplication cysts reported, which were resected only by endoscopy in adults. In pediatric surgery there is only one case reported with a tubular type esophageal duplication cyst that was treated by endoscopic resection. The case is about an 11 year old boy who presented with chest pain, dysphagia, cough and fever. The endoscopic examination showed a double lumen of the esophagus, which was resected by a diathermic knife in the same session. The patient had an uneventful postoperative course and could be discharged at the third postoperative day. The follow-up 18 month later was also uneventful.

The presented case is the second in literature reporting about an endoscopic resection in children and the first case using the insulated scissors- type knife SB knife (SUMITOMO BAKELITE CO., Tokyo, Japan). Compared with former therapeutic strategies based on laparoscopic approach the endoscopic treatment could lead to early begin with oral nutrition and to shorter hospital stay.

#### 4. Conclusion

Endoscopic resection could be a save treatment option for a tubular type esophageal duplication cyst also in children. Compared to other therapeutic approaches the advantages are less invasiveness, less complications and shorter hospital stay. Due to

the rare number of patients more cases and more experience are necessary to establish the endoscopic approach as state of the art therapy for tubular type esophageal duplication cysts.

#### Acknowledgments

Thanks to my husband and family for their support.

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